

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Living with pulmonary hypertension: unique insights from an international ethnographic study
AUTHORS	Kingman, Martha; Hinzmann, Barbara; Sweet, Oliver; Vachiéry, Jean-Luc

VERSION 1 - REVIEW

REVIEWER	Laura Savage VCU Medical Center USA
REVIEW RETURNED	21-Feb-2014

GENERAL COMMENTS	Excellent paper- unique methodology. A few comments: at the end of the introduction section, stated "we aimed to include patients with PAH or CTEPH"- why were these separated out? CTEPH is part of PH classification- you included subjects from Class 1 and Class 4? Under methods, what are "patient associations"? This was not clear to the reader. Also under methods, it was stated "patients had access to the identity of the sponsor". Is this a requirement to state this? RE: Data collection-- how did the researcher film subjects-- was it a stationary camera or did a researcher actually follow the subject around for 6 hrs? Did the diaries have a guide or was it open to pt interpretation? Can you more fully describe what "projective and creative techniques" were used? Under Analysis section: how was the 45 min ethnographic film derived? What was the "single 1 hour cross country film" This section should be more fully delineated. Under Personality and daily routine, recommended describing each category more fully and using pt quotes to support your statements.
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REVIEWER	Silvia Ulrich University Hospital of Zurich, Switzerland
REVIEW RETURNED	10-Mar-2014

GENERAL COMMENTS	This is a very interesting study on the patients' perspective and perception of their disease "pulmonary hypertension" (PH). As a PH-specialist dealing daily with affected patients, this study provides important insights from the other side, the patients with their environment, feelings, sorrows, fears and handicaps in their daily life. I am sure that these insights will be important for many caregivers and therefore warrants publication. As University teacher in Medicine, I am familiar with scientific studies dealing with numbers, graphs and measures leading to statistics with predefined significance levels. The method used here is a qualitative one, ethnography, derived from social sciences, which I'm not familiar in.
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	<p>However, having performed several studies mainly in the field of PH, I exactly know that evidence-based medicine with randomized trials itself has many drawbacks which sometimes makes transfer to everyday life difficult, e.g. patients eligible for studies are highly selected and do not necessarily represent the broad collective seen in the clinic and that endpoints used in trials are not necessarily the once that are meaningful for patients in their lives. Quality of life (QoL) has therefore emerged as an endpoint and questionnaires were validated by research groups, two of them by our (Minnesota living with heart failure questionnaire, Cenedese E. et al, ERJ 2006, CAMPHOR in German, Cima K, Health and QoL Outcomes 2012). However, questionnaires deal with various aspects of the disease, but are not individualized and thus do not give the holistic perspective as given by qualitative research. Therefore, the authors have to be congratulated to perform this task and share their results with healthcare providers in PH but potentially also patients and their organisation. I would appreciate if this kind of research would continue to show differences between countries, age groups and PH-classes, potentially subgroups or settings. The paper is well written and understandable.</p>
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VERSION 1 – AUTHOR RESPONSE

Reviewer: 1

Excellent paper- unique methodology.

We would like to thank the reviewer for her comments, which we have addressed point-by-point below.

A few comments: at the end of the introduction section, stated "we aimed to include patients with PAH or CTEPH"- why were these separated out? CTEPH is part of PH classification- you included subjects from Class 1 and Class 4?

The study focussed on patients with PAH and CTEPH as the most recognized forms of PH, and the only forms for which approved treatments were available at the time of study start (PAH-specific pharmacotherapies and pulmonary endarterectomy, respectively). Furthermore, many patients with CTEPH also receive off-label treatment with PAH-specific pharmacotherapies and indeed, this was part of the inclusion criteria for entry of patients with CTEPH into the study. We therefore felt that it was important to include these patients only, and not those from classes 2, 3 and 5 whose treatment is more distinct from patients with PAH and CTEPH.

Under methods, what are "patient associations"? This was not clear to the reader.

The term 'patient associations' refers to organisations such as the Pulmonary Hypertension Association that provide a number of services to the pulmonary hypertension community, including support, education, research and advocacy. In certain countries, such as South Korea, recruitment of patients into our study via physicians was impossible due to strict local guidelines and regulations. Thus, recruitment had to occur through local patient associations. To clarify this we have amended the manuscript to read 'patient associations (organisations that provide a number of services to the PH community, including support, education, research and advocacy)'.

Also under methods, it was stated "patients had access to the identity of the sponsor". Is this a

requirement to state this?

It is a requirement in research such as this to disclose the name of the sponsor at the end of the interview, should the patient ask. It was not stipulated by the journal that this information should be provided in the manuscript. However, we feel that it is important to be as transparent as possible when disclosing the role of the sponsor in the study.

RE: Data collection-- how did the researcher film subjects-- was it a stationary camera or did a researcher actually follow the subject around for 6 hrs?

The researchers followed the patients for 6 hours with a camera. Filming was conducted in an empathetic manner, with a small hand-held camera. We have amended the methods section to reflect this.

Did the diaries have a guide or was it open to pt interpretation? Can you more fully describe what "projective and creative techniques" were used?

The patients were not provided with a guide for completing the diaries. The diaries had open questions, offering the opportunity for a qualitative interpretation of the patients' experiences of living with PAH and CTEPH. The projective and creative techniques refer to the way the diaries were structured into four written tasks titled: my perfect day, how my treatment makes me feel, letter to my doctor and if my illness was a person. Projective/creative techniques are research tools or approaches designed to access thoughts, feelings or needs that are not easily accessible to research participants and/or to the researcher. These techniques offer a structure for research participants that makes it easier for them to access thoughts and emotions that are difficult to verbalize or difficult to express publicly.

Under Analysis section: how was the 45 min ethnographic film derived?

The 45 minute ethnographic film was derived following analysis sessions, which included the ethnographers in the field, the analysts at the research agency headquarters in London who used transcripts and fieldnotes, and healthcare experts within the research agency. We have added further details to the methods regarding this process.

What was the "single 1 hour cross country film" This section should be more fully delineated.

The single 1-hour cross-country film highlighting the key findings across the countries was produced and edited thematically to allow cross-cultural comparison. We have added this extra detail to the manuscript.

Under Personality and daily routine, recommended describing each category more fully and using pt quotes to support your statements.

As requested by the reviewer, we have provided more detailed descriptions of each category and have added further patient quotes in support of our statements.

Reviewer: 2

This is a very interesting study on the patients' perspective and perception of their disease "pulmonary hypertension" (PH). As a PH-specialist dealing daily with affected patients, this study provides important insights from the other side, the patients with their environment, feelings, sorrows, fears and handicaps in their daily live. I am sure that these insights will be important for many

caregivers and therefore warrants publication. As University teacher in Medicine, I am familiar with scientific studies dealing with numbers, graphs and measures leading to statistics with predefined significance levels. The method used here is a qualitative one, ethnography, derived from social sciences, which I'm not familiar in.

However, having performed several studies mainly in the field of PH, I exactly know that evidence-based medicine with randomized trials itself has many drawbacks which sometimes makes transfer to everyday life difficult, e.g. patients eligible for studies are highly selected and do not necessarily represent the broad collective seen in the clinic and that endpoints used in trials are not necessarily the once that are meaningful for patients in their lives. Quality of life (QoL) has therefore emerged as an endpoint and questionnaires were validated by research groups, two of them by our (Minnesota living with heart failure questionnaire, Cenedese E. et al, ERJ 2006, CAMPHOR in German, Cima K, Health and QoL Outcomes 2012). However, questionnaires deal with various aspects of the disease, but are not individualized and thus do not give the holistic perspective as given by qualitative research. Therefore, the authors have to be congratulated to perform this task and share their results with healthcare providers in PH but potentially also patients and their organisation. I would appreciate if this kind of research would continue to show differences between countries, age groups and PH-classes, potentially subgroups or settings. The paper is well written and understandable.

We would like to thank the reviewer for her comments. We completely agree that future research should investigate differences between countries, age groups and PH-classes, and potentially subgroups or settings. Unfortunately this was not possible in the current study due to the small sample size, a limitation that we have acknowledged in the discussion.

VERSION 2 – REVIEW

REVIEWER	Laura Savage, RN, MSN, CCTC VCU Medical Center USA
REVIEW RETURNED	14-Apr-2014

GENERAL COMMENTS	Excellent paper. Important work to understand the qualitative nature of pt's experiences.
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